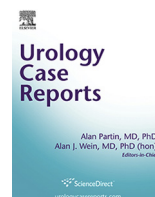


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## Urology Case Reports

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## Inflammation and Infection

## Xanthogranulomatous Cystitis Treated by Transurethral Resection

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## ABSTRACT

Xanthogranulomatous cystitis (XC) is a rare benign chronic inflammatory disease of unknown etiology. Curative treatment of XC requires surgical resection, and most of reported cases were treated by partial cystectomy. Here we describe a case with XC that was treated using transurethral resection.

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## Introduction

Xanthogranulomatous changes, histologically characterized by the presence of lipid-laden macrophages, multinucleated giant cells, and cholesterol clefts, have been reported in many sites,<sup>1</sup> and the kidney is the most common site in the urinary tract. Xanthogranulomatous cystitis (XC) is a rare benign chronic inflammatory disease of unknown etiology. In this report, we describe a case with XC that was treated using transurethral resection.

## Case presentation

A 64-year-old female presented with repeated cystitis. At presentation, she had no lower urinary tract symptoms or hematuria. She had a history of a pyelolithotomy for a right renal stone and a partial ureterectomy with an ureterovesicostomy for a right megaureter at the age of 30. Furthermore, she had a hysterectomy at the age of 50 and a laparoscopic cholecystectomy in the previous year. Physical examination was unremarkable, and laboratory data revealed a normal hematological and biochemical profile. Urinalysis revealed 0–5 red blood cells and 0–1 white blood cell per high power field. Urine culture grew coagulase-negative *Staphylococcus*,  $\alpha$ -hemolytic *Streptococcus*, and *Corynebacterium* species. Urinary cytology showed no malignant cells. Ultrasonography revealed mild hydronephrosis in the right kidney, bilateral multiple renal

cysts, and thickening of posterior bladder wall with no blood flow (Fig. 1). Her voiding function was normal with negligible post-void residual urine. Cystoscopy revealed a papillary mucosa at the posterior wall (Fig. 2). Enhanced computed tomography demonstrated no stone or tumor in the right ureter and no appreciable change in the bladder. Magnetic resonance imaging revealed no urachal remnants. Cystoradiogram did not show vesicoureteral reflux.

Because the presence of bladder neoplasm could not be ruled out, an endoscopic resection was performed. Histological examination revealed a yellow-brown specimen, including many foamy macrophages that stained positive for CD68 (Fig. 3), infiltrated neutrophils, and fibrosis in the submucosal layer. These histological findings led to the diagnosis of XC. There was no evidence of malignancy. The postoperative course was uneventful. The patient had no recurrence 10 months after the surgery.

## Discussion

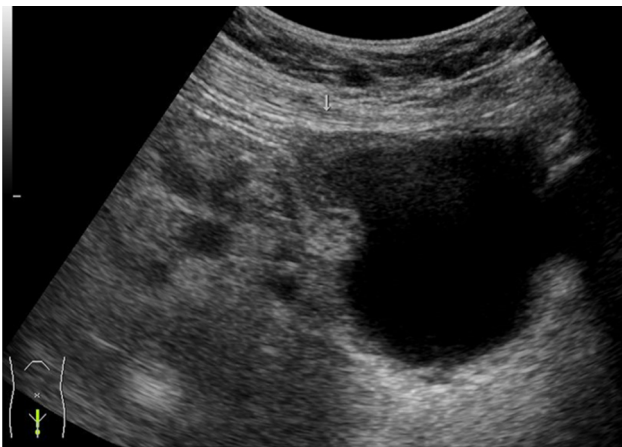
XC is a rare inflammatory disease that is microscopically characterized by multinucleated giant cells, lipid-laden macrophages (xanthoma cells), and cholesterol crystals, and macroscopically by soft yellow-brown plaques.<sup>1</sup> Since the first report by Wassiljew in 1932,<sup>2</sup> 27 cases of XC have been reported in the literature. All 28 cases, including the present report, had the following clinical characteristics<sup>1–5</sup> (Table 1): a median age of 46 years (range, 16–76 years) and no sexual predominance (14 cases in males, 13 cases in females, and one unknown).

The pathogenesis of XC is unclear. In the previously reported cases, the proposed etiology includes an urachal remnant, a chronic infection, foreign materials such as retained suture materials, a

Abbreviations: XC, xanthogranulomatous cystitis.

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**Figure 1.** Ultrasonic image of a sagittal view of the bladder showing thickening of the bladder wall with no blood flow.

malignant bladder tumor, immunological disorders, abnormal lipid metabolism, and urethral stenosis.<sup>1,3–5</sup> In the kidney, xanthogranulomatous inflammation always develops as a consequence of an infection, which is often associated with urinary obstruction. In the present case, the XC may have been caused by repeated cystitis or retained suture materials.

The location of XC was at the dome of the bladder in 18 of 28 cases, and 14 of the 28 cases were associated with an urachal lesion (Table 1). In the present case, the posterior wall of the bladder was involved, which indicates that there was no relationship with an urachus.

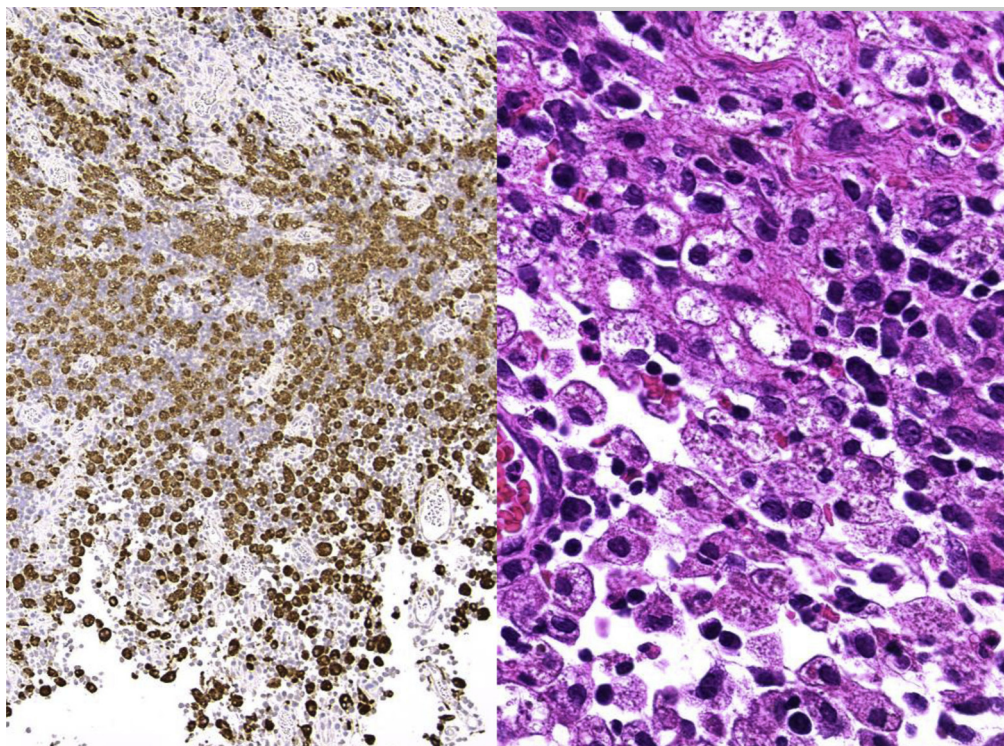
XC has no specific clinical manifestations; therefore, it is difficult to distinguish from other bladder diseases, such as carcinoma. The



**Figure 2.** Cystoscopy showing edematous papillary mucosa at the posterior wall.

main symptoms were lower abdominal palpable mass, cystitis-like symptoms, discharge from the umbilicus, and painless macro-hematuria. Therefore, the diagnosis of XC is usually based on pathological examinations.

Curative treatment of XC requires surgical resection,<sup>3</sup> and no postoperative recurrence has been reported. Of the 28 published cases, the treatment measure was partial resection in 22 cases, transurethral resection in four cases including the present case,



**Figure 3.** Microscopic examination of the lesion showing the presence of many foamy macrophages that stained positive for CD68 (left, ×100). Hematoxylin and eosin staining is shown in the right panel (×400).

**Table 1**  
Characteristics of the reported XC cases

Age	Mean 46 (16–76)
Sex	
Male	14
Female	13
Unknown	1
Site	
Dome	18
Posterior wall	3
Lateral wall	2
Vesico-ureteric junction	1
Associated pathology	
Urachal remnant	11
Urachal adenoma	2
Urachal adenocarcinoma	1
Ulcerative colitis	2
Urothelial carcinoma	1
Symptom	
Abdominal mass	9
Frequency	9
Dysuria	7
Lower abdominal pain	7
Hematuria	6
Umbilical pus	3
Urgency	3
Treatment	
Partial cystectomy	22
Total cystectomy	1
Augmentation cystoplasty	1
Transurethral resection	4

total cystectomy in one case, and augmentation cystoplasty for a small capacity bladder after the exclusion of neoplasm in one case. The use of transurethral resection to treat XC was reported only in

recent years,<sup>5</sup> possibly because of early detection via imaging tests. Transurethral resection appears to be a minimally invasive and curative treatment for small XC lesions.

## Conclusion

XC is a rare benign chronic inflammatory disease. Curative treatment of XC requires surgical resection, and a partial cystectomy has been applied in most cases. The present case suggests that a transurethral resection is a minimally invasive and curative treatment for small XC lesions.

## Conflict of interest

The authors of this manuscript have no conflict of interests to disclose.

## Consent

The patient has provided permission to publish the features of her case.

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